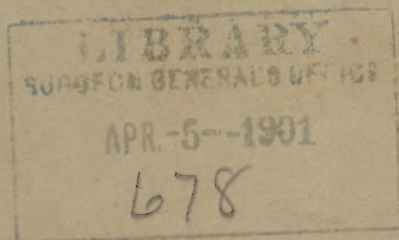


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REPORT OF
TWO CASES OF ERYTHROMELALGIA
(*Red Neuralgia*—MITCHELL).

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*Read before the
Association of American Physicians,
May 4, 1897.*



TWO CASES OF ERYTHROMELALGIA (MITCHELL).

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CASE I. *Female, aged fifty-eight years; left hand only affected.*—Mrs. C., aged fifty-eight years; neurotic diathesis, but has kept up and worked hard in household duties, keeping a summer hotel. Before this attack was “run down” physically. Disease first began in August, 1895, with burning pain in first finger of left hand at angle of nail next the thumb, and continued for two weeks in the first finger. In the autumn of 1896 extended to second and third fingers, then to the fourth, and finally to the thumb.

A large, tender swelling developed at the base of the thumb, and this winter there is another tender spot on the back of the hand, over the third and fourth metacarpal bones. Sometimes over the abductor minimi digiti muscle. The pain is of a severe, burning character, lasting sometimes for days together, with remission, but not entirely absent during the attack.

The ulnar artery was cut at the wrist several years ago by a broken lamp-chimney, and was tied. The patient thought, perhaps, the nerve was injured, and this caused the trouble. This is hardly likely, however, as the pain began and has been most severe in the radial side. Stimulating the ulnar nerve at the elbow causes twitching of the third and fourth fingers.

The character of the pain during an exacerbation is of a severe, burning nature, and paroxysmal, but the parts are always tender until the attacks pass off, then there will be freedom from pain until the next attack. Previous treatment consisted of local applications, as chloroform, tincture of iodine, blistering, etc., and general treatment in the form of tonics, and especially for dyspepsia.

Paroxysms of pain were in recurring attacks from three to six weeks apart. Pain was better on lying down. Had typhoid fever thirty years ago. No history of malaria or of specific disease.

October 31, 1896. She came under my care, suffering from the symptoms just stated. The pain was evidently very severe, as shown by the facies. It was felt most severely in the thumb and first and second fingers of the left hand, which were of a purplish-red hue and swollen. The worst pain was in the ball of the thumb, which was swollen and very tender. The general health was impaired. Heart's action weak, but otherwise normal. She was

very nervous and slept badly on account of the pain. There was also pain up the arm to the back of the neck—described as a drawing pain, extending also to the ears, jaws, and throat, with a dull ache of the head and blurring before the eyes.

No dizziness, but occasional difficulty in walking (ataxia). Knee-jerk normal.

Dyspeptic symptoms very marked, and aside from the pain in the hand it was the dyspepsia which called especially for treatment.

The character of the indigestion is that common in neurasthenia—from innervation—not from organic stomach disease.

For relief of pain :

R.—Phenacetin	2.00
Caffeine	1.00

M.—Ten capsules. S.—Two every hour until relieved or until six are taken.

Usually one dose gave partial relief.

She was given nitroglycerin, one and one-half milligramme, three times a day, with tonics and bismuth and carefully regulated diet. After two weeks of the treatment without benefit, galvanism was applied from the hand to the back of the neck daily—three minutes at a time. This was continued with the nitroglycerin and tonics for a month, without apparent relief, the daily pain still continuing. It was then stopped for awhile, but the pain became worse, and galvanism was again resorted to. The relief, however, was nothing to boast of, though it appeared to be the treatment that offered the best hope of good result.

The nitroglycerin to strengthen the action of the weak heart and stimulate freer passage of blood through the capillary system, by dilating arterioles, also electricity, the constant current, with positive pole in the hand and the negative at the back of the neck, should relieve pain in the hand and relax spasm.

The treatment here outlined was carried out for three months with little or no relief. The patient improved somewhat in general health, but there was no improvement of the pain in the hand. It fluctuated—sometimes better, sometimes worse—until

February 18th, when the most violent attack of all occurred, the pain being of an intense, burning character, extending to the whole hand, though more severe at the points previously indicated, and following up the arm to the shoulder.

The color was purplish-red with œdematous swelling of the parts most affected. The last joint of the middle finger was livid, as from strangulation, and appeared as though gangrene might follow.

During the previous night she had taken six capsules of phenacetin and caffeine (of the former 1.20 grammes in all) without relief of pain, but with profuse sweating.

A prescription containing extract of *cannabis indica*, 0.03; extract of

FIG. 1.



Palm of hand during a paroxysm; Feb. 18, areas of pain shown by the shading.

FIG. 2.



Back of hand during a paroxysm; Feb. 18, areas of pain shown by the shading.

belladonna, 0.01, and salicylate of cinchonidia, 0.20 to the dose, to be taken every three hours, was ordered, and at night hypodermic.

Holding up the hands gives relief.

A hypodermic injection of morphia and atropia was given, which later secured relief and a good night's rest.

FIG. 3.



Palm of hand showing character of the swelling.

Pain returned with less severity the following day.

The hypodermic was repeated in a smaller dose the next night, with the same good effect, and then no longer needed.

CASE II. *Female, aged thirty-seven years. Disease in both feet.*—Mrs. A., widow, aged thirty-seven years, a neurasthenic, with severe dyspeptic symptoms. No constitutional taint, as far as known; no history of malaria. Pain in the heels began three or four years ago, and has recurred at intervals of three or four months, the attacks lasting from two to three weeks. During the interval is comparatively free from pain.

The paroxysms seem to be brought on by physical exhaustion or any kind of strain to the nervous system, especially if obliged to stand or walk more than usual, as on one occasion when called to nurse her mother in a prolonged illness.

The pain begins as a dull ache with burning sensation; at first with a sensation of pebbles under the heels. As the attack progresses the burning increases and extends to the sole of the feet and up the calves of the legs.

When pain becomes severe the heels are red and swollen, and shoes cannot

be worn, as the attack increases in severity. A peculiar sensation extends to the head, restlessness and hysterical symptoms, and a disposition to scream. When asked as to severity of pain, said it was so bad that she felt "she just could not stand it another moment."

Lying down relieves, except when at the worst, then the restlessness forced her out of bed, and rubbing would give her temporary relief. Cold relieves. Putting the feet out from under the cover was grateful. At my suggestion cold water was tried with benefit. Attacks more frequent and worse in summer than in winter and also when feet get warm in bed.

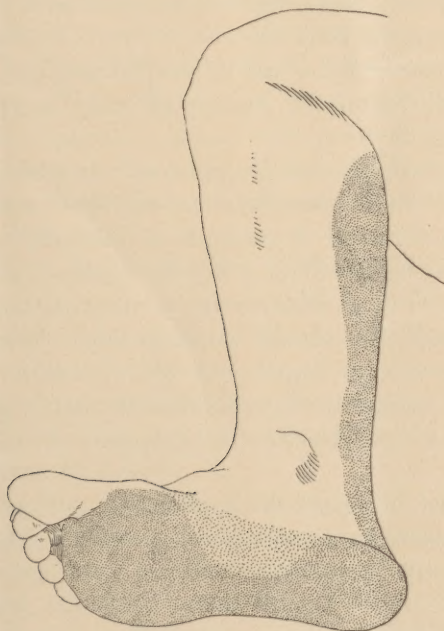
She is drowsy and dizzy much of the time, and frequently shows an ataxic gait.

Treatment has been directed to the general condition—the dyspepsia and neurasthenic symptoms.

She has an idiosyncrasy against strychnia, the latter causing dizziness and vertigo.

No disease of special organs discoverable.

FIG. 4.



Shaded areas show location of pain.

This disease is undoubtedly a rare one, and fortunately so, for, as Dr. Mitchell says, treatment is almost unavailing, though patients

often go on for years without becoming worse. It occurs almost always in the feet. Out of sixteen cases reported by Dr. Weir Mitchell (*American Journal of the Medical Sciences*, 1878, vol. lxxvi.) only one was in the hands alone, and in two the hands and feet were both affected. All of the others were in the feet. He gives as the characteristic symptoms redness, intense burning pain, and at the beginning rise of temperature, but later fall of temperature, with probably inflammation of nerves affected. It usually commences in the ball of the foot, or the heel, or the great toe, and extends to the sole and dorsum. The disease is more common in men than in women—twelve out of the sixteen cases reported by Dr. Mitchell were men.

Another peculiarity of this affection is that it is aggravated by heat and relieved by cold. In Dr. Mitchell's cases relief came from lying down, although in a case treated by prolonged rest—while the patient was perfectly comfortable in bed—the pain returned in full force on getting up and walking.

In the first case cold does not relieve the pain, and but little relief accompanies the recumbent posture, probably because it is in the hands instead of the feet.

The causes of the disease are obscure. It seems to follow a depressed state of the nervous system, neglected neurasthenia, long standing on the feet, long marches, as of soldiers; reflex irritation, especially of the male urethra. But the disease is so rare and these causes so common that such reference is unsatisfactory.

As to treatment, it is equally unsatisfactory. The indications are to relieve symptoms and improve the general health.

Neurotic or neurasthenic symptoms by baths, spinal douches, good food and air and tonics. The dyspepsia should receive attention.

For the relief of pain, cold applications. These cases are, as a rule, made worse by heat and relieved by cold. They are worse in summer and better in winter, so that a cool summer climate is indicated.

Galvanism—the positive electrode at the periphery; the negative to back of neck.

Rest in bed, where pain is very severe.

Possible surgical operation where indications are found. In the

present case I think stretching or cutting of the nerves of the forearm should be tried if the pain continues.

In regard to medicine to relieve the pain, Case I. has had phenacetin and caffeine with good effect. Cannabis indica and belladonna proved unavailing.

From the chronic character of the pain opium and its preparations should be avoided if possible. But sometimes the suffering is of such an intense character that morphia is imperatively called for, preferably under the skin in combination with atropia.

As to diagnosis in erythromelalgia, according to Dr. Mitchell, there is no other disease or group of symptoms in a well-marked case with which it can be confounded. There are many painful affections of the feet and hands, such as the effects of gout, rheumatism, sprain from flat-foot, nodes from syphilis, etc. ; but none in which the train of symptoms found here exist. And that the disease is rare is evident from the comparatively few cases reported, although Dr. Mitchell predicts that attention having been called to this form of nerve disease it will be found more common than previously supposed. The pathology of these cases is involved in doubt. Erythromelalgia is classed among the vasomotor diseases, such as angio-neurotic oedema and Raynaud's disease. The pain either precedes or is simultaneous with the redness and swelling. In the case of Mrs. C. they came on at the same time. In the second case pain preceded the redness. This would seem to indicate that the sensory fibres of the nerves were first affected, the vasomotor becoming involved secondarily as a result. This is also borne out by the severe pain on pressure at the points most affected. With the redness and swelling, however, the pain is greatly aggravated.

The diseases to which it is most nearly allied are angio-neurotic oedema and Raynaud's disease.

In the former there are three forms described. The pale form (neuro-lymphangioma), due to dilatation of the lymphatics of paretic origin ; the red form, due to angio-paralysis, and the dark-red, purplish form, caused, according to Eskridge (Wilson's *Applied Therapeutics*, p. 981), by spasm of the arterioles, which "checks the *vis a tergo* of the arterial current in propelling the current forward, and spasm of the small veins, which may force the blood backward into the arterioles."

Some such explanation as this may apply to erythromelalgia, but is altogether hypothetical.

Angio-neurotic œdema is supposed to be a form of urticaria—"the greater urticaria"—a functional nervous disease, caused by indigestion or transient nervous excitement. In some of the cases which I have seen it has been attributed to a spider bite—I need not say without cause. The element of pain is secondary in angio-neurotic œdema, and not important.

This disease cannot be confounded with the one under consideration.

The diagnosis for Raynaud's disease, though still more marked, deserves special notice, since many of the cases of erythromelalgia reported by Mitchell were pronounced by high authority to be cases of Raynaud's disease (Osler's *Practice of Medicine*, 1892, p. 962). The two diseases in many respects are the opposite of each other. This is perhaps best shown in the comparison in parallel columns given by Lannois (article on "Erythromelalgia," 1880, p. 71; Weir Mitchell's *Clinical Lessons on Nervous Diseases*, 1897, p. 179).

Local asphyxia (Raynaud).

Sex, four-fifths females.

Begins with ischæmia.

Affected part becomes bloodless and white. In certain cases there is the deep, dusky congestion of a cyanosed part, with or without gangrene.

Pain may be absent or acute, and comes and goes; has no relation to position; may precede local asphyxia.

Unaffected by seasons. In many cases all the symptoms can be brought on by cold.

Anæsthesia to touch.

Analgesia.

Temperature much lowered and unaltered by posture.

Gangrene local and limited; likely to be symmetrical.

Erythromelalgia (Mitchell).

In twenty-seven cases two were females.

Little or no difference in color until foot hangs down in upright posture, when it becomes rose-red.

Arteries throb and parts become of a dusky red or violaceous in tint.

Pain usually present; worse when part hangs down or is pressed upon. In bad cases more or less at all times.

Worse in summer and from heat; eased by cold.

Sensation of all kinds preserved.

Hyperalgesia.

Temperature greatly above normal; dependency causes, in some cases, increase of heat; in others lowering of temperature.

No gangrene; asymmetrical.

This comparative statement should make it clear that erythromelalgia and Raynaud's disease are distinctly separate.

Two other conditions are mentioned as belonging to diseases of the vasomotor system, namely, adiposis dolorosa (Dercum's disease)

and acromegaly, both attended with peculiar changes of nutrition and without doubt from disease of the central nervous system.

In acromegaly there was found "enlarged pituitary body, with enlarged sella turcica, persistence of the thymus gland, and hypertrophy of the fibres and glands of the vasomotor system (Wilson: *American Text-book of Applied Therapeutics*, 1896, p. 988). These facts are mentioned as having a bearing upon the plausibility of erythromelalgia being essentially a central disease.

There is more reason for this hypothesis in the fact that a number of the cases reported developed into well-marked disease of the cerebro-spinal axis and ran their course to a fatal result (Mitchell). Dr. Mitchell was disposed at first to this opinion, but in his recent work (*Clinical Notes*, 1897, p. 180) he "inclines rather to some of that new clinical perplexity, peripheral neuritis."

The action of ergot (ergotism) in producing gangrene of the extremities is of interest as bearing upon the general question of causation of vasomotor affections by central disease.

Ergot acts upon the vasomotor centres—in small doses by stimulating, in large doses by depressing (H. C. Wood's *Therapeutics*). Dr. H. C. Wood mentions a case (*loc. cit.*) of fatal ergot-poisoning in which there were gastric irritation, thirst, diarrhœa, *burning pain* in the feet, and convulsions.

The principal vasomotor centre is in the medulla oblongata, between the corpora quadrigemina and calamus scriptorum (Ludwig and Thiry, Owsjannkow; and Dittmar, Erb-Dis. Medulla Oblong.). Also in spinal cord, secondary centres in gray matter as far down as the lumbar vertebræ (Goltz, Vulpian, *loc. cit.*).

Dr. Schenk, in an article published recently in the *Medicinische Presse*, states the disease is no doubt due to vasomotor disturbance consequent on an ascending degeneration in the posterior column of the cord. Woodnut has also described a case of erythromelalgia, attributing it to myelitis (London *Lancet*).

Referring again to Mitchell's *Clinical Notes*, he says:

"At the time I wrote (1878) I should, if driven to be positive in statement, have inclined toward considering this malady as due to some form of spinal disorder.

"But at present the reasonable explanations incline rather toward some form of that new clinical perplexity—peripheral neuritis." (*Loc. cit.*, p. 180.)

In Case LVIII. (of *Clinical Lessons*, not the fifty-eighth case of erythromelalgia) the disease was the result of an injury—a stone falling on the foot.

This case is reported in great detail, and serves to strengthen the theory of peripheral neuritis, from the fact that it was caused by (or followed) a local injury. Dr. Mitchell further goes on to say :

“ I use the word neuritis, nerve-end neuritis, with more or less doubt. Some such distinct affection of the smaller nerves does seem to me probable, but whether it is congestion, neuritis, or some other of the undescribed changes in the lesser nerves or in the ultimate nerve-plates, we may not as yet decide with certainty.”

Again (*loc. cit.*), “ Neuritis is becoming a sad puzzle. We may have it with paresis and little pain; we may have it without notable paresis and horrible pain. It exists with or without notable myositis.”

Again, “ It may give rise to causalgia, joint-troubles, and alterations of nails and hair. It may fail to disturb nutrition, or greatly to alter local heat. And, lastly, if erythromelalgia be a neuritis it may cause pain and flushing, and to these increased enormously by dependency of the part may add such a rise of temperature as is rarely seen in acute local inflammation.”

Dr. Mitchell continues to discuss the possibility of the existence of neuritis of such a character as to single out individual elements in a nerve containing fibres of sensation, motion, nutrition, and vasal control—affecting some and not others. Such an hypothesis seems untenable.

In Case LVIII., previously referred to, the excised portions of the musculo-cutaneous and internal saphenous nerves were examined microscopically and found “ absolutely normal.”

To sum up the pathology of them, we have the choice between :

1. Disease of the central nervous system, or
2. Peripheral neuritis affecting principally or only the smaller nerves or nerve-endings.

In the former we should expect the disease to be located in the medulla oblongata between the corpora quadrigemina and calamus scriptorum, or in the posterior segment of the spinal cord as far down as the lumbar vertebræ.

The central theory seems the more plausible, but at this time is purely hypothetical, there being no dissections to support it.

The opinion of peripheral neuritis of a peculiar character is given

a show of plausibility from two cases reported by Dr. Mitchell which followed local injury.

However, in both of these cases operated on, the nerves were exsected, and the nerve-trunks thus removed found normal.

This fact would locate the neuritis in the nerve extremities.

We will close this report by a reference to treatment, which, unfortunately, can be shortly disposed of. Thus far curative treatment has been of no avail, except in one case, by surgical interference.

The treatment divides itself into three indications:

1. That of the general health—neurasthenia, nervous dyspepsia, and hysteria especially.

2. The relief of pain. For this purpose phenacetin protected by caffeine seems of first value. Cannabis indica and belladonna in the first of the present cases proved useless, as did also the phenacetin in the most violent paroxysms.

During the latter subcutaneous injection of morphia is imperative, but the objection to its frequent use is obvious. Rest in bed and cold applications have their place. Wendel reports a case cured by liq. acidi arseniosi, together with general treatment (*New York Medical Record*, 1890, vol. xxxviii. p. 545).

3. Surgical treatment. Exsection or stretching of the nerves was contemplated in the first case, but there being no precedent for it, and fearing serious nutritive changes, it was not determined on. This was before reading Dr. Mitchell's *Clinical Lessons*, 1897, in which he reports two cases thus treated, the first one with success in relieving the patient. In the second case, however, gangrene, resulting fatally, followed.

The operation in the former cases, the first of its kind in this disease, I quote from Dr. Mitchell's book (*loc. cit.*, p. 192):

"Dr. Keen operated on April 10th. He exsected two and a half inches of the muscular cutaneous nerve and the same length of two branches of the internal saphenous. The two end branches (plantar) of the posterior tibial nerve were stretched at the internal malleolus with a traction of fifteen pounds thrice used. The result next day was remarkable. There was almost immediate relief. The foot could be squeezed, pinched, or pricked without pain. Anæsthesia was not as extensive as we were led to expect it might be. The day following the operation the temperature—right, 95°; left, 93.5°; mouth, 99.5° (it was the right foot operated). On the right clonus still present, and knee-jerk was still extensive. The wound healed in a

few days. On May 4th the patient was able to walk on crutches. Clonus had disappeared and reflexes were no longer exaggerated. There was still some flushing when pendant, but no pain nor hyperæsthesia. When discharged—May 6th—he could walk well, but was ordered to continue the use of crutches as a measure of precaution, and had also a bandage applied daily.

“ Dr. Kyle reports culture of blood and nerves obtained at the operation as yielding purely negative results. . . . June 7th, S. is reported well, and walking easily without crutches. Six months later he was at his work, as a stonemason, and entirely free from pain.”

Thus it appears that at least one was cured by surgical procedure. The second case thus treated (Case LIX. of *Clinical Lessons*) was more unfortunate. I will only mention the operation and result.

Operation April 12, 1894, by Dr. T. P. Morton. Four inches of the musculo-cutaneous nerve and five inches of the internal saphenous excised. The posterior tibial behind the internal malleolus stretched with force of twenty-eight pounds. On the fifth day gangrene developed. A week later amputation was performed, with death on the operating-table.

Commenting on this case, Dr. Mitchell says: “As to operative relief I think that in future I should stretch all the nerves and leave the section of their trunks to be resorted to if milder means gave no fortunate results” (*loc. cit.*, p. 204).

In conclusion I append titles of bibliography of erythromelalgia, taken from the card catalogue of the Library of the Surgeon-General's Office. These titles do not appear in the large published *Index Catalogue*.

The disease had not been differentiated at the time the volume of the *Index Catalogue*, in which they would have appeared, was compiled.

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